# Temporary Hidden Aneurysms During Pregnancy

# A Case Report

C.S. CHO, Y.J. KIM, K.T. CHO, S.K. LEE, B.J. PARK, M.K. CHO

Department of Neurosurgery, Dankook University College of Medicine, Cheonan; Korea

Key words: pregnancy, brain aneurysms, subarachnoid haemorrhage

## **Summary**

There are a number of reports on cerebral aneurysmal ruptures during pregnancy. Although the cerebral aneurysmal rupture is quite rare during pregnancy, it leads to a high maternal mortality; which gives rise to a clinical significance.

We have encountered a number of multiple cerebral aneurysms during pregnancy, and the ruptured cases were successfully treated with the coiling procedures. The coiling is found to be a good treatment method for ruptured aneurysms during pregnancy.

The cerebral angiogram is the gold standard diagnostic method for detecting cerebral aneurysms. However, 1.8-20% of the cases are reported to be false negative in initial angiography, and only up to 20% can be diagnosed even with the additional angiographies. A special attention for a hidden aneurysm should also be required while managing the patients.

#### Introduction

The incidence of intracranial haemorrhage from a rupture of cerebral aneurysm is quite rare during pregnancy <sup>1,2</sup>, but the mortality rate of maternal death due to the aneurysmal rupture is as high as 5 to 12% of all its causes <sup>1,2</sup>. Particularly, a rupture of intracranial aneurysm

during pregnancy may lead to a fatal result not only to the maternal body but also to the fetus. Therefore, it is difficult to decide the timing of treatment and its relevant methods<sup>3</sup>. A surgical clipping has been the most commonly used treatment method. However, since the introduction of Gugulemi Detachable Coil (GDC), its applications are being increasingly used owing to the development of instruments for the endovascular coiling and the radiological apparatuses<sup>4</sup>.

During the coil embolization process of a patient with multiple intracranial aneurysms during pregnancy, the authors experienced a relatively large hidden aneurysm. We report the case together with a literature review.

# Case

A 26-year-old woman who was in the 36<sup>th</sup> week of gestation visited our institution due to symptoms of acute headache and decreased consciousness. Hunt and Hess (H-H) grade of the patient was three and the volume of haemorrhage was measured Fisher Grade 3. (figure 1A). On the first day in the hospital, an aneurysm at the right dominant anterior communicating artery (AcomA) was found in cerebral angiography (figure 1B), and the coiling was implemented under Monitored Anesthesia Care (M.A.C) by using Propofol and Alfentanyl.

R







Figure 1 A) Initial noncontrast brain CT scan. There are hyperdense subarachnoid haemorrhage in the category of Fisher grade 3. B) Right internal carotid angiography reveals aneurysm of the right dominant anterior communicating artery aneurysm. C) Right internal carotid angiography shows complete occlusion of anterior communication artery aneurysm.

The patient's abdomen was covered with a lead apron with a purpose of protecting the fetus. After the aneurysmal coiling (figure 1C), cesarean operation was performed; the baby was in a good condition.

The patient showed a stable progress until a development of a sudden cardiac arrest on the 12<sup>th</sup> day of the hospitalization. Soon after the cardiopulmonary resuscitation was done, neurological changes including left mydriasis, III, VI N palsy, and diplopia, etc. developed. As a

result, computerized tomography and cerebral angiography was followed, but a rebleeding, other particular symptoms, nor any abnormalities in blood sample was not found. However, another aneurysm at the basilar artery (BA) was observed on the cerebral angiography (figures 2A,B). Hence an additional coiling was done in the aneurysm (figure 2C). The patient was discharged from the hospital without any neurological defects on the 30th day of the hospitalization.

# Discussion

An intracranial haemorrhage from a rupture of cerebral aneurysm is a lesion that shows a high rate of mortality and morbidity. Although the incidence of cerebrovascular diseases during pregnancy is still not quite clear, an approximation of 0.3 to nine patients per 100,000 persons<sup>5</sup> was reported in some studies. The reports described its causes in connection with changes in hormones with haemodynamic aspects during pregnancy<sup>2</sup>.

A subarachnoid haemorrhage from a cerebral artery aneurysmal rupture has been the 3<sup>rd</sup>

most common non-obstetric cause during pregnancy <sup>2,6</sup>. A high incidence rate can be observed in the third trimester. According to the report by Hunt et Al <sup>6</sup>; about 6% of rupture of cerebral artery aneurysm occurred in the first trimester, about 31% in the second trimester, about 55% in the third trimester, and 8% after delivery.

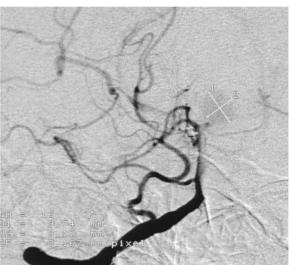
The rates of rebleeding and maternal deaths are also high. Referring to some reports, in the case of rebleeding, the maternal death rate was measured up to 50-68% <sup>2</sup>. Because it displays such a high rate of maternal death, a careful treatment considering both maternal body and fetus should be required. Recently, the efficacy and the safety of aneurysmal coiling have been reported in a number of reports <sup>4</sup>.

There have been many discussions and controversies concerning the relationship between a pregnancy and a cerebral haemorrhage 7,8. Some argue that there is no relation of a pregnancy to a cerebral haemorrhage 7; however, in more recent report, its relationship with a pregnancy has been asserted 9. It is thought that changes in haemodynamics and hormones play major roles in the formation of cerebral aneurysms during pregancy. The increase of the blood volume reaches to a maximum level of approximately 1700 ml in around 32nd week of gestation, which leads to an increase of 25-40% comparing to the one before pregnancy; it consequently leads to an increase of the cardiac output. On the aspect of changes in hormones; the hormones such as estrogen, progesterone and prostacyclin affect the vascular smooth muscle. As a result, this affect the vascular resistance to decrease 10.

In cases with neurological deficits including sudden acute headaches and decreased consciousness state, it is necessary to differentiate it from pituitary apoplexy, intraarterial occlusions, dural sinus thrombosis, intracranial space-occupying lesions, meningitis, encephalitis, demyelinating diseases, eclampsia, and etc<sup>2</sup>. As for the method of diagnosis, lumbar puncture, CT and MRI, etc. can be considered.

Figure 2 A) Right vertebral artery angiography shows no abnormal findings except for some vasospasm at admission. B) Follow-up. The vertebral angiography showing an aneurysm of the basilar artery on hospital 12 days. C) Right vertebral artery angiography shows complete occlusion of basilar artery aneurysm.







In considering the effects of the irradiation, MRI is recommended when the state of patient allows, especially in the first trimester 11. However, the radiation exposure on the fetus in the later stage in pregnancy is allowed up to maximum of 0.5 rem. When CT is used and a lead apron, etc. are installed over the uterus, the exposure to the radiation is measured less than 0.05 rem; and with the performance of angiography, less than 0.1 rem is exposured 12. It has been reported that a contrast agent has no effect on fetus; however, when using it over dose, fetus may show a transient hypothyroism. The fetus should also be protected from dehydration when using a contrast agent<sup>2</sup>. As previously stated, when exposing to an ionized radiation, it is recommended that a lead apron to be used. Mannitol, an osmotic diuretic, may induce hypovolemia and hypotension in maternal body, which may lead to the uterine hypoperfusion, and consequently the fetal hypernatremia and hyperosmolarity<sup>2</sup>.

The teratogenic effect of anticonvulsants during the first trimester has been reported, and they lie in Category D of the U.S. Food and Drug Administration. Carbamazepine, which is in Category C, is known to be the most favorable among the anticonvulsants <sup>13</sup>. Since hyperventilation is a potential cause of acid-base shifts and decreased oxygen delivery to the fetus, the fetal heart rate should be monitored for signs of fetal distress <sup>8</sup>.

In cases of cerebral aneurysms in the basilar artery, a patient may easily experience neurological changes by a mass effect due to the location of the aneurysm being near the brain stem and the cranial nerves in the anatomic aspect. The symptomatic aneurysms are closely related to the rapid growth of an aneurysm; Sekhar and et Al <sup>14</sup>, described it with the axial stream impingement and the water hammer effect, etc.

The cerebral angiogram is the gold standard diagnostic method for detecting cerebral aneurysms. However, 1.8-20% of the cases are reported to be false negative in initial angiography, and only up to 20% can be diagnosed even with the additional angiographies <sup>15</sup>. In spontaneous SAH patients with negative CT findings, Rinkel et Al <sup>16</sup>, suggested that an additional angiography should be followed if there is a diffuse distribution of perimesencephalic haemorrhage; an additional angiography is recommen-

ded only in cases with a focal haemorrhage. Inamasu et Al<sup>17</sup>, reported that an anterior communicating artery is the most common location of the cerebral aneurysms in patients with the initial false negative CT finding and the positive additional angiographic result. They insisted on the necessity for a close attention around the site; it is reported that even a surgical exploration was carried out in angiogram-negative patients. The reason for such a high false negative rate around the A-com complex is believed that the size of the aneurysms might be too small for detection and the blood flow might find very difficult to enter the aneurysms due to the turbulence <sup>17,18</sup>.

The incidence of rebleeding is significantly lower in patients with negative angiograms than in patients with ruptured aneurysms; the reports show the rate around 0-5%19.

Although a number of theories are proposed, the etiology of SAH with negative angiographic results still remains not clear. Some theories suggest that the microaneurysms may be destroyed or thrombosed when the rupture occurs <sup>20</sup>. Rinkel et Al <sup>16</sup>. made a proposition of the role of venous or capillary source in perimesencephalic SAH patients. On the other hand, Alexander et Al <sup>21</sup> suggested the leakage of the lenticulostriate and thalamoperforating vessels as the predominant cause for the SAH.

Other reasons have also been reported, and they are vasospasm, spontaneous thrombosis of the aneurysm, arterial dissection, haematoma, and tamponade effects <sup>22</sup>.

In addition, the authors believe that high contrast injection rate and amount, aneurysms with small orifice, high growth rate of the aneurysms, missed interpretation of the imaging study results, and physicians' unawareness of the hidden aneurysms could affect the cerebral aneurysmal rupture.

## **Conclusions**

The method and timing of the treatment is important in ruptured intracranial aneurysms during pregnancy. Aneurysmal coiling is a good treatment option for ruptured aneurysms during pregnancy. When a hidden aneurysm may be discovered by additional examinations, a few different reasons could be suspected. The authors recommend to always keep in mind of possible existence of hidden aneurysms.

#### References

- 1 Barno A, Freeman DW: Maternal deaths due to spontaneous subarachnoid haemorrhage. Am J Obstet Gynecol 125: 384-392, 1976.
- 2 Dias M, Sekhar LN: Intracranial haemorrhage from aneurysms and arteriovenous malformations during pregnancy and the puerperium. Neurosurgery 27: 855-865, 1990.
- 3 Ingall TJ, Whisnant JP et Al: Has there been a decline in subarachnoid haemorrhage mortality? Stroke 20: 718-724, 1989.
- 4 Molyneux A, Kerr R et Al: International Aneurysm Trial (ISAT) of neurosurgical clipping versus endovascular coiling in 2143 patients with ruptured intracranial aneurysms: a randomised trial. Lancet 360: 1267-1274, 2002.
- 5 Shutter LA, Kline LB, Fisher WS: Visual loss and a suprasellar mass complicated by pregnancy. Surv Ophthalmol 38: 63-69,1993.
- 6 Hunt HB, Schifrin BS, Suzuki K: Ruptured berry aneury and pregnancy. Obstet Gynecol 43: 827-837, 1974
- 7 Kittner SJ, Stern BJ et Al: Pregnancy and the risk of stroke. N Engl J Med 335: 768-774, 1996.
- 8 van Buul BJ, Ninjhuis JG et Al: General anesthesia for surgical repair of intracranial aneurysm in pregnancy: Effect on fetal heart rate. Am J Perinatol 10: 183-186, 1003
- 9 Shutter LA, Kline LB, Fisher WS: Visual loss and a suprasellar mass complicated by pregnancy. Surv Ophthalmol 38: 63-69, 1993.
   10 Ortiz O, Voelker J, Eneorji F: Transient enlargement of
- 10 Ortiz O, Voelker J, Eneorji F: Transient enlargement of an intracranial aneurysm during pregnancy: Case report. Surg Neurol 47: 527-531, 1997.
- 11 Kopitnik TA, Samson DS: Management of subarachnoid haemorrhage. J Neurol Neurosug Psychiatry 56: 947-959, 1993.
- 12 Piper J: Fetal toxicity of common neurosurgical drugs. In Loftus C (ed): Neurosurgical Aspects of Pregnancy. Park Ridge, IL, American Association of Neurological Surgeons 1-20, 1996.
- 13 Saunders M: Épilepsy in women of childbearing age. BMJ 299: 581, 1989.
- 14 Sekhar LN, Heros RC: Origin, growth, and rupture of saccular aneurysms: a review. Neurosurgery 8: 248-260, 1081
- 15 Suzuki S, Kayama T et Al: Subarachnoid haemorrhage of unknown cause. Neurosurgery 21: 310-313, 1987.

- 16 Rinkel GJE, Wijdicks EFM et Al: Nonaneurysmal perimesencephalic subarachnoid haemorrhage: CT and MR patterns that differ from aneurysmal rupture. Am J Neuroradiol 12: 829-834, 1991.
- 17 Inagawa H, Wakai S et Al: Ruptured cerebral aneurysms missed by initial angiographic study. Neurosurgery 27: 45-51, 1990.
  18 DiLorenzo N, Guidetti G: Anterior communicating
- 18 DiLorenzo N, Guidetti G: Anterior communicating aneurysm missed at angiography: report of two cases treated surgically. Neurosurgery 23: 494-499, 1988.
- 19 Nishioka H, Torner JC et Al: Cooperative study of intracranial aneurysms and subarachnoid haemorrhage: a long-term prognostic study. III. Subarachnoid haemorrhage of undetermined etiology. Arch Neurol 41: 1147-1151, 1984.
- 20 Hayward RD: Subarachnoid haemorrhage of unknown etiology. A clinical and radiological study of 51cases. J Neurol Neurosurg Psychiatry 40: 926-931, 1977.
- 21 Alexander MSM, Dias PS, Uttley D: Spontaneous subarachnoid haemorrhage and negative cerebral panangiography. Review of 140 cases. J Neurosurg 64: 537-542, 1986.
- 22 Tatter SB, Crowell RM, Ogilvy CS: Aneurysmal and microaneurysmal "angiogram-negative" subarachnoid haemorrhage. Neurosurgery 37: 48-55, 1995.

Young-Joon Kim, M.D. Ph.D. Department of Neurosurgery Dankook University College of Medicine 29-Anseo-dong, Cheonan, Choongnam 330-714, Korea E-mail: kimyj@dku.edu